

Submandibular Gland Sialolith in a Renal Transplant Recipient: A Case Report

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Abstract

Salivary dysfunction may be due to systemic diseases and medications. The development of sialoliths is a multifactorial event in which disturbances in secretion, microliths, and bacteria may play a major role. A case of sialolith in the submandibular gland of a 58-year old woman, with a medical history of renal failure end kidney transplant, is reported.

Keywords: Sialolith, submandibular gland, kidney transplant

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Introduction

Sialolithiasis is a common disease of the salivary glands and is caused by formation of sialoliths.¹ Sialoliths are calcified masses that develop in the intra- or extra-glandular duct system and form as a result of mineralization of debris that have accumulated in the duct lumen. These debris may include mucous plugs, bacterial colonies, exfoliated duct epithelial cells, foreign bodies, or other cellular debris.² Chemically, sialoliths are condensations of calcium salts, primarily calcium phosphate and calcium carbonate showing the apatite structure, with small amounts of other inorganic and organic components.³

Sialolithiasis may occur in any major salivary gland but is most common in the submandibular gland.¹ There are two reasons for this phenomenon. The submandibular saliva is rich in mucus and is, thus, more viscous than parotid saliva. In addition the submandibular duct ascends when the body is upright, it bends at the posterior edge of the mouth, and its course is long and sinuous.⁵ Because of the horseshoe-shape of the body of the gland, this duct has a bow-shaped course in the cranial direction, which means there is a particular tendency in this gland to secretory congestion and concrement formation.

Histologically, sialoliths have a concentric, laminated structure of alternating layers of organic and inorganic substances. The calculi are often built up around one or more central cores, while in some cases a central core is lacking.⁶

The precise cause of sialolith formation is still largely unexplained but infection or inflammation of the gland, the viscous nature of mucous secretions, and others have all been suggested as predisposing factors for their development.⁴

The disturbance in salivary secretion and the change in the composition of saliva, which was called dyschylia by Seifert, leads to an increase in salivary viscosity and to a slime obstruction in the terminal ducts of the gland. The disturbance in salivary secretion and increased formation of microliths in ducts support the ascent of bacteria and cause focal obstructive atrophy of the gland parenchyma.⁴ Salivary dysfunction may be due

to systemic diseases, medications, and head and neck radiotherapy.⁸ Loss of renal function results in the accumulation of metabolic waste products and alters the normal hemostatic mechanisms that control fluid and electrolyte balance.¹⁰ It is described that many uremic patients suffer from xerostomia.⁹

This paper presents a case of sialolith of the submandibular gland in a renal transplant patient.

Case Report

A 58-year old woman was referred to the São Lucas Hospital (PUCRS, Brazil) with the chief complaint of a painful swelling on the left submandibular gland, accompanied by difficulty in swallowing. She had recurrent episodes of swelling and pain of the submandibular region over the past few years. On anamnesis she revealed she has been on dialysis in the past, had a kidney transplanted the last year, and was under immunosuppressive therapy. On physical examination, the patient had a purulent discharge from the left Wharton duct with a palpably enlarged submandibular gland. Ultrasonography and a radiograph suggested the existence of an intraglandular sialolith (Figures 1 and 2).

The diagnosis of sialolithiasis with suppurative inflammatory was made. After antibiotic and anti-inflammatory treatment, the patient was admitted into the hospital. Under general anesthesia, a large calculi and the left submandibular gland were removed using a Risdon approach (Figures 3 and 4).

The Wharton's duct was closed by an intra-oral approach (Figure 5). The histopathological diagnosis of the removed specimen was chronic sialadenitis caused by sialolithiasis. The surgically removed submandibular gland contained an intraglandular sialolith with normal structure in a dilated duct (Figure 6). A thickened layer of fibrous connective tissue with chronic inflammation surrounded the sialolith. At 3 years' follow-up, there has been no clinical sign or symptom of recurrence.

Discussion

Sialolithiasis is the most common disease of the salivary glands in middle-aged patients. It



Figure 1: Panoramic radiograph shows a large radiopaque mass in the left mandibular area.

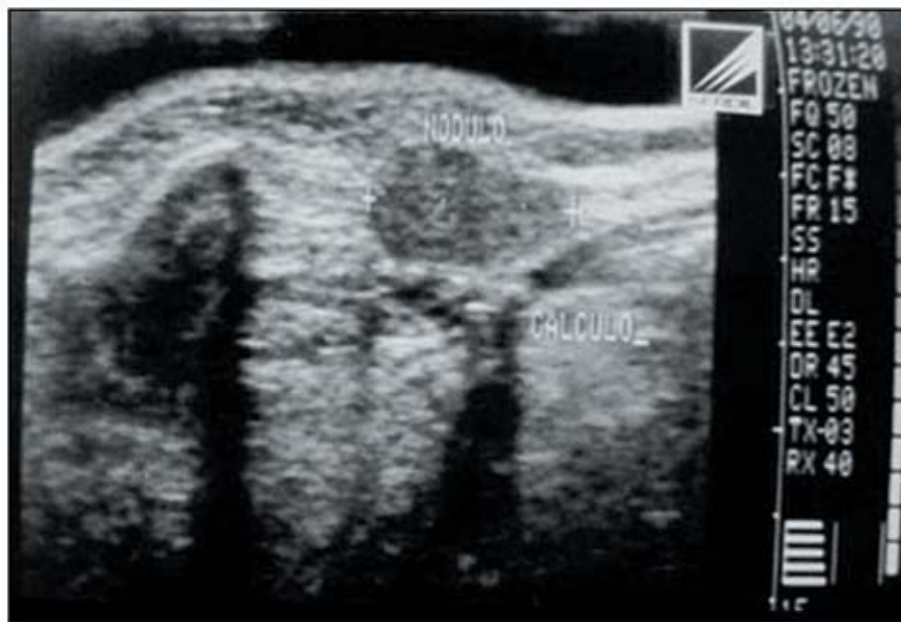


Figure 2: Ultrasound view of the right submandibular gland in showing stone.



Figure 3: Risdon approach for excision of submandibular gland.



Figure 4: Gross specimen of the left sublingual gland and thesialoliths.



Figure 5: Closure of Wharton's duct by interior approach.

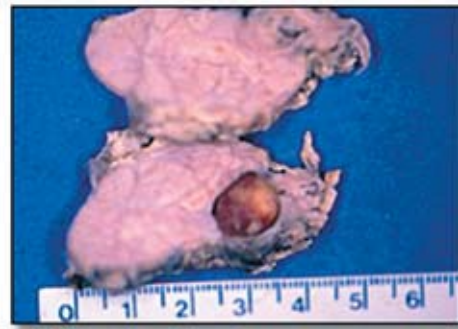


Figure 6: Internal view of removed sublingual gland with irregular submandibular sialolith, measuring 10 mm.

is estimated sialolithiasis affects 1.2% of the population.¹¹ More than 90% of the sialoliths occur in the submandibular gland.¹ The great majority of these sialoliths are localized in the Wharton's duct. The ratio of sialoliths localized intraglandularly to those in Wharton's duct is 3: 7.¹¹ Compared to the other salivary glands, the submandibular gland is rich in viscous secretion of high alkalinity. A further difference is the length of the excretory duct.

Sialoliths can occasionally be palpated using bidigital palpation techniques in the floor of the mouth (for submandibular and sublingual calculi) and parotid regions. Sialograms can identify changes in the salivary gland architecture and are useful for major salivary gland swellings.¹² They are performed with radio-opaque iodide and extraoral radiographs (lateral cephalograms, panoramographs). Ultrasound and computerized tomography (CT) scans will help rule out salivary gland tumors and sialoliths.⁸

Despite the very extensive literature, the pathogenesis of salivary calculi still remains unclear.² Salivary calculi grow by deposition, at a rate that has been estimated at approximately 1–1.5 mm per year, and range in size from 0.1 to 30 mm.¹³

As the patient grows older and consumes more medication, in many cases, reduced secretory activity, alterations of electrolyte concentrations, impairment of glycoprotein synthesis, and structural deterioration of cell membranes of the salivary glands are seen.^{15, 16} All of these may contribute to the underlying mechanisms for the higher incidence of calculi among the elderly.

The most common types of medications causing salivary dysfunction have anticholinergic effects via inhibition of acetylcholine binding to muscarinic receptors on the acinar cells. Any drug that inhibits neurotransmitter binding to acinar membrane receptors or that perturb ion transport pathways, may also adversely affect the quality and quantity of salivary output. These medications include tricyclic antidepressants, sedatives and tranquilizers, antihistamines, and antihypertensives. Therefore, patients taking one or more drugs with antisialogogue sequelae should be followed carefully for developing signs and symptoms of salivary disorders.¹⁴

A prominent feature of the present case was the fact the elderly patient reported previous xerostomia and had been taking antihypertensives since the renal failure. After transplantation, the patient will remain on one or more immunosuppressive agents for the rest of his or her life to prevent organ graft rejection. The most commonly used drugs include cyclosporin, azathioprine, and steroids.¹⁷ Cyclosporin can cause severe renal damage with secondary hypertension.

Loss of renal function results in the accumulation of metabolic waste products and alters the normal hemostatic mechanisms that control water and electrolyte balance.¹⁰ When the disease progresses and the failing kidneys worsen, medical management alone cannot prevent azotemia. At this point, there are two treatment alternatives to prolong life: artificial filtration of blood by means of dialysis or transplantation of a kidney.¹⁸

Oral lesions related to renal disease are generally nonspecific. However, many of the metabolic and physiologic body alterations that accompany renal disease have oral manifestations.¹⁹ Elevation of blood urea nitrogen in renal failure results in a high concentration of urea in the saliva.^{19, 20} It has been suggested the cause of stomatitis in uremic patients is partially related to the high salivary urea level with consequent breakdown into ammonia and harmful metabolites, which are not being executed by the kidneys.¹⁹ Oral lesions are considered to be a result of mucosal irritation by ammonia, which is released by the action of urease-containing microorganisms on salivary urea.²⁰ It is well known most uremic patients suffer from cutaneous dryness and skin desquamation of various degrees. Xerostomia has also been described in these patients.²¹ The desquamation of the epithelium may be related to the increased urea content of saliva, mucosal irritation by ammonia, electrolyte disturbances, and the surface dryness of gingiva due to the existing xerostomia.¹⁹⁻²¹

The macro and microstructure of the hydroxyapatite salivary calculi is practically identical to those found in the hydroxyapatite renal calculi (non-infective phosphate renal calculi).²² The structural similarities between both types of calculi must also involve a similar mechanism of formation. Thus, similar to the hydroxyapatite renal calculi, small particles of organic matter are the initial substrate on

which the calculus development starts. An interesting parallelism between the formation of these calculi and the renal calculi exists. In both cases the presence of retained organic matter, a high hydroxyapatite supersaturation, and the deficit of crystallization inhibitors would permit the development of the first spherulites of hydroxyapatite in the organic matrix, which upon causing a still more effective obstruction of the salivary duct favors the calculus growth.²³

Conclusion

It can be concluded the etiologic factors implied in the sialolith formation can be classified into two large groups: (a) saliva retention due to morphoanatomic factors (salivary duct stenosis, salivary duct diverticuli, etc.) and (b) saliva composition factors (high supersaturation, crystallization inhibitors deficit, etc.). Obviously, the existence of a bacterial infection can favor the development of sialoliths through the increase of salivary pH and due to the increase of organic matter that can obstruct the salivary ducts.

The presence of renal disease or systemic diseases that alter the salivary function may be a predisposing feature for salivary calculi formation. In this particular case the sialolith in the submandibular gland seems to be multifactorial and associated to the patient's medical history of renal failure end kidney transplant.

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